VAN: HOOK (W.)

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BY

WELLER VAN HOOK, A.B., M.D., Professor of Surgery in the Chicago Polyclinic.

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STRANGULATED INGUINAL HERNIA OF A CYSTIC APPENDIX VERMIFORMIS.*

By Weller Van Hook, A. B., M. D., Professor of Surgery in the Chicago Polyclinic.

Leopold M., German by birth, forty-three years old; weight, one hundred and seventy pounds; height, five feet nine inches; newspaper dealer; had had good health (except an attack of typhoid fever at fourteen years of age) up to four years ago. At that time he had a feeling of soreness at the umbilical region, and a sudden movement, a concussion or pressure upon the abdomen, would cause intense pain. As a result of overexertion in lifting, a right oblique inguinal scrotal hernia was contracted a year and a half ago. The abdominal tenderness gradually increased up to the time I first saw him. He had never suffered from indigestion or griping pains. He has always been constipated, the bowels sometimes not moving for a week. He never noticed a tumefaction in the abdomen, but on pressure over the iliac and hypogastric regions he felt a sensation of tenderness. This soreness was not greater upon one side than upon the other. The hernia always disappeared without taxis on lying down, until September 16. 1895, when he noticed that the hernial contents remained in the sac. His own efforts to reduce it being unsuccessful, Dr. J. W. Dal was called after thirty-six hours. Dr. Dal found a tense swelling in the inguinal canal which he could not reduce without anæsthesia. The patient was vomiting fluids from the upper intestinal tract. No gas had passed by the rectum, but this fact may have been due to lack of sufficient peristaltic activity. Under chloroform Dr. Dal again attempted taxis, and the tumor seemed to disappear. When the patient recovered from the anæsthetic, however, pain was greater than before. and took on a different character, radiating over the lower abdomen toward the umbilicus. The doctor's observation at the evening visit convinced him that the patient's condition was not satisfactory, and a

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consultation was requested. On examining the patient, I found him suffering from some abdominal pain, with pulse of 90°, and a slight elevation of temperature. There was tenderness over the lower right side of the abdomen. In this region the muscles were abnormally rigid and palpation was difficult. The abdomen was but slightly distended. In the inguinal canal and extending into the scrotum was a brawny mass, not very tense. It seemed most reasonable to suppose, in the absence of clearly marked diagnostic signs, that reduction had been imperfectly accomplished, and that a thin mass of omentum was left in the sac, together with a knuckle of intestine.

After the usual preparations, the sac was exposed by a sufficient incision and carefully opened. To my surprise, no omentum or intestine was present. The brawny sensation was due to acute inflammation of an unusually thick sac. The peritoneal lining of the sac was injected and rough and the walls ædematous. In the sac, lying in scattered masses here and there, was a quantity of translucent ambercolored mucus having almost exactly the appearance of calf's-foot jelly broken up into pieces of one to ten millimetres' thickness. Its consistence, however, was somewhat tenacious. Having seen on another occasion the same material in a cyst of the vermiform appendix, I recognized at once that such a cyst had been strangulated in the inguinal canal and ruptured in reduction.

The incision was prolonged sufficiently to open the abdominal cavity, where an additional quantity of mucus was found scattered over the intestines and the parietal peritonæum of the internal iliac fossa sufficient to make the whole amount equal to at least an ounce. The caput coli was near at hand, and on drawing it into the wound the cystic appendix was drawn into the wound. At the distal extremity of the appendix was found a rent as large as a lead pencil, through which mucus was protruding. The walls of the appendix were dilated at the middle, but near the cæcum was a constriction. The caput coli and neighboring small intestines were covered with thick tenacious masses of false membrane, quantities of which were stripped off. There was but a small amount of fluid present. The peritonæum, where not covered with false membrane, was deeply injected and roughened. That these changes involved an extensive area of the serous membrane was proved by cautiously drawing out the neighboring coils of bowel.

The sac of the hernia was first dissected free and cut off at the coeliotomy wound, a number of iodoform-gauze capillary drains were applied carefully in different directions in the abdomen, and the caput

coli drawn well up to the wound. The appendix was then ligated close to its base and amputated with scissors. The suture used to ligate the appendix was left long enough to be passed through the eve of a needle, and was then carried through the muscular walls of the abdomen. In this way the cæcum, whose walls were more violently inflamed than those of other parts of the intestines, was anchored close to the abdominal wound. The coeliotomy opening was, for the most part, left open to facilitate drainage, although a few silkwormgut stitches were inserted, to be tightened as secondary sutures. The patient bore the operation well, and, despite the grave local infection, made uninterrupted progress toward recovery. During the first few days an enormous serous discharge escaped from the opening, and the temperature went up to 101.2°. The pain disappeared, however, and, the bowels responding to laxatives, the general condition of the patient rapidly improved, the wound healed by granulation, and the man now, three months after the operation, attends to his usual business, wearing a truss to protect the abdominal walls.

The portion of the appendix removed is, in its recent condition, six centimetres in length and two centimetres and a half in diameter. Its outer (peritoneal) surface is of a bright-red color except at points not covered by a grayish-red false membrane. Here and there are small masses of an amber-colored, thick, tenacious substance which finds its way out of the appendix, on pressing its walls, through the cut end and through an irregular opening at the distal extremity about eight millimetres in diameter. The shape of the appendix is irregularly cylindrical; but about two centimetres from the distal extremity the tube is bent upon itself from the longitudinal axis in the direction of the mesenteriolum at an angle of about 40°. The walls of the appendix vary in thickness from about four millimetres at the point of amputation to nine or ten millimetres near the distal extremity. There are also variations in the thickness of the walls at the same level, giving rise to slight pouchings of the lining membrane at several points. But these pockets do not at any point reach the dignity of diverticula. The interior of the appendix is filled with the same gelatinous, amber-colored material already noted as having been seen adherent to the peritoneal surface of the process.

Microscopically, the fibrous coats of the appendix are greatly thickened. The muscular layers are not thicker than normal. Out of thirty sections examined, only one shows a small, ill-developed layer of epithelial cells representing the atrophic mucous membrane, the place of which elsewhere is filled entirely by a layer of connective tissue. This case is unique in the occurrence of rupture of the cystic appendix in the sac of a hernia; it is the third case of which I have knowledge, after a study of the accessible literature, where a cystic dilatation of the appendix occurred in a hernia, and the ninety-ninth case of hernia of the appendix. It is also interesting from the fact that inflammation of a hernial sac had its origin in a lesion of the appendix.

Rupture of the cystic appendix by taxis in the effort to reduce an inguinal hernia is an accident which, as has been already remarked, has not before been recorded. And even if one were aware of such a possibility no practical advantage of the knowledge could be taken, both on account of the rarity of the circumstance and the want of diagnostic signs to lead us to a proper knowledge of the conditions before operating. The practical lesson to be gained from an acquaintance with this case is that the contents of a hernia are practically unknowable before the sac is opened, and that taxis is dangerous in a certain proportion of cases. But little persuasion is needed to convince surgeons to-day that the open method is, in the great majority of instances, the simplest, most radical, and satisfactory treatment for strangulated and incarcerated hernias.

Cystic enlargement of the vermiform appendix seems first to have been noted by Virchow, who had observed a case in which the appendix was distended to the size of a man's fist, and as the contents were a thin watery fluid, he called the condition "hydrops of the vermiform appendix."

Rokitansky, in his *Text-book of Pathological Anatomy*, said: "We further occasionally observe a metamorphosis of the vermicular process produced by obturation, which is analogous to dropsy of the efferent ducts of glands, and which is most apparent in the gall bladder (hydrops cystidis felleæ)."

Glasmacher's case was that of a soldier who presented himself with a tumor the size of a pigeon's egg in the right inguinal canal near the abdominal ring. For a year there was no trouble. Suddenly obstruction occurred, and on making a herniotomy a cyst holding a drachm of pus was found. This cyst contained no fæces or gas. A pedicle extended into the abdominal cavity. The patient died with continued symptoms of obstruction. At the autopsy intestinal gangrene was found. The cyst was proved to have been the appendix. It does not seem to have been a true retention cyst.

Zdekauer's case was that of a woman, eighteen years of age, who had a recent right inguinal incarcerated hernia the contents of which

were diagnosticated as an ovary. On exposing the mass it was found to be a vermiform appendix dilated to the size of an acorn and filled with pus. This case, like that of Glasmacher, I can not regard as a retention cyst on the evidence of the recorded histories, although Bajardi classifies them in that category. The condition was simply that of pus retention within the imprisoned appendix.

Wölfler's case was operated upon by Billroth for hernia. No intestine was found, but a cyst, thought at first to be the sac, was opened, allowing a teacupful of serous fluid to escape. The herniated cyst was found to communicate with a larger cystic cavity within the abdomen, extending from the psoas muscle to the small pelvis. Pressure on the abdomen caused a bloody fluid to escape. It was thought the symptoms of strangulation had been brought about by the tenseness of the cyst. In spite of free drainage, the patient did not do well until the lining membrane of the cyst, after several days, had sloughed and been removed with forceps. The thickness of the cyst wall was between three and seven millimetres.

Steiner describes four specimens in the pathological museum of Basle, giving chief attention to minute anatomy:

Case I was from the body of a woman, thirty years of age, who died of puerperal fever. The contents of the cyst were homogeneous gelatinous masses. No communication existed between the cyst and the cæcum. The form of the cyst was elongated, biscuitlike, two dilated parts communicating by a narrow isthmus. The entire length of the appendix was 5.5 centimetres. The thickness of the wall varied between one and 2.5 millimetres. The inner surface of the cyst showed a shining smooth membrane with striated markings. No trace of epithelium or Lieberkuehn's glands. Even the muscularis mucosæ and the follicles had disappeared. The wall of the cyst consisted of three layers, an inner connective tissue layer, and a layer of loose cellular tissue, uniting the first layer with the well-developed muscular layer, consisting chiefly of circular fibres.

CASE II.—The appendix was 8.8 centimetres long. The cystic dilatation had a longitudinal extent of 5.8 centimetres, with a transverse diameter of 1.8 centimetres.

CASE III was also that of a small cyst. The mucous membrane was not wholly destroyed.

CASE IV occurred in the person of a woman fifty-five years old. The cyst was small and the muscularis mucosæ, together with some epithelium, was retained *in situ*.

Steiner states that in 2,280 protocols of autopsies made in Basle,

only three cysts were found. In the cases with moderate dilatation a distinct hypertrophy was found in the circular fibers of the muscular layer, while in the larger cysts, with marked stretching of the wall, the musculature was not thickened. Steiner, like other observers, noted the occurrence of diverticula in the walls of the cysts. His discussion of the microscopical relations of the walls of the cysts is interesting, but too long to be quoted.

Shoemaker published an account of a case in which a cystic dilatation of the appendix was discovered post mortem, no symptoms having been noted during life.

The six post-mortem specimens of Ribbert had fortunately such varying characteristics that he was able to formulate the changes which take place in the appendix when cystic dilatation occurs. He says:

"In the lesser degrees (of dilatation) we see no essential alteration in the composition of the wall. Later, abnormities of the epithelium occur. It is lost, together with the glands, throughout considerable areas, and forms in other parts a covering of a single layer without glands, which, however, may still be present in patches. In very great dilatation it is destroyed altogether, but, as Case VI shows, disappears even when dilatation is slight. We must, however, take into consideration that the loss of epithelium does not need to be the result of dilatation, but that it may be destroyed by the diseased process which led to the cicatrization producing the obliteration of the lumen. The follicles disappear equally soon, and finally are altogether wanting."

Ribbert says that an obliteration interrupting the lumen of the tube does not necessarily lead to a cystic dilatation, although the glands may be retained. Bischoff thinks that a dilatation is wanting when the mucous membrane is still able to resorb in the normal way. Ribbert thinks a dilatation is also wanting when, as a result of early and extensive destruction of the mucous membrane, no secretion into the lumen can longer occur.

Finkelstein describes a specimen in Sonnenburg's collection in which the appendix was dilated to a length of fourteen centimetres and a circumference of twenty-one centimetres. The pear-shaped appendix appeared like a pedunculated tumor.

In the case reported by Wenzel-Gruber was a cyst six centimetres long attached to the cæcum by a pedicle four centimetres long. The cyst had a transverse diameter of four centimetres and a half, and was filled with viscid mucus. This case was remarkable for the fact that

the obliteration of the lumen was brought about by a chronic tubercular catarrh.

Kelynack relates a case of a middle-aged female who died from extensive vegetative endocarditis, in which the appendix was found to be completely shut off from the cæcum, and no sign or indication of any previous communication could be observed. The appendix was greatly distended and presented two very distinct diverticular processes, which were directed between the folds of the mesentery of the appendix. The diverticula were connected with the dilated cavity of



FIG. 1.—Kelynack's case of cystic dilatation of the appendix. "The cystic organ has been laid open in its greater part. A portion of the anterior wall of the cæcum has been removed in order to show the proximal occluded end of the appendix bulging into it."

the appendix through well-defined circular openings. The appendix contained a thick, gelatinous, light-yellow substance, and also a small quantity of material like curdled milk in appearance.

J. A. Berry refers to a case of Féré described as a mucocele of the appendix, and Berry himself describes a case in which (as in Shoemaker's case) the appendix was found post mortem distended with thick gelatinous mucus.

Orth and Leube give descriptions corresponding with the older accounts of Virchow and Rokitansky.

Bierhoff gives a report of three cases of this condition.

Foerster is cited by Woelfler as having had a case.

Kelynack, in addition to his own case, refers to reports by Fenwick and Coats. The latter states that in his case the appendix had been converted into a large cyst, measuring five inches in its long diameter. The cyst contained a tenacious colored material, and the wall was thick and firm. In Fenwick's case "the appendix was distended by a milky fluid, the communication with the cæcum being obliterated."

In February, 1895, I witnessed an operation by Mr. Frederick Treves at the London Hospital in which the appendix was enlarged cylindrically, its length appearing to be about four inches and a half and its diameter one inch. What symptoms had occurred to indicate an operation were not stated. The appendix was amputated after the formation of cufflike flaps which were united over the stump. The cyst contained a thick gelatinous mucus. Mr. Treves stated that a specimen almost exactly similar to the one removed was deposited in the Cambridge Museum. Mr. Treves' case is the only one operated on for symptoms arising in a cyst not involved in a hernia. A record of the symptoms produced would be of great interest.

A most interesting case is that of Maylard, who found at a post mortem upon a patient dying of Bright's disease a dilated appendix measuring four inches in length and two inches in thickness. The dilated part was filled with a clear gelatinous substance which could be turned out en masse. At the proximal end it communicated freely with the cæcum. Perhaps we would be less surprised at the presence of so much mucus if we remembered that it is not uncommon to find a viscid plug of mucus in the normal appendix. And as Maylard's case seems well authenticated, we must assume that absolute obstruction is not wholly necessary to the accumulation of thick mucus. It must be otherwise when the fluid is limpid, as in Virchow's and Rokitansky's hydropic form. As in my case the base of the appendix was ligated before being removed, no attempt was made to determine the permeability of the proximal part of the process, but the occurrence of active plastic peritonitis strongly indicates the partial patency of the tube, permitting the exit of micro-organisms from the cæcum. The contents of such cysts containing thick mucus can not therefore be regarded as sterile without further investigation. It would seem probable that micro-organisms were absent from the cysts whose contents are limpid. In the case of the cysts whose contents are viscid and gelatinous, the stiff and unyielding character of the material accounts for its failure to be expelled through the contracted opening. while the more fluid secretions could easily escape.

Prof. Ludwig Hektoen has kindly placed at my disposal a specimen of cystic dilatation of the appendix removed from the body of a

woman who had died from nephritis. The appendix is about seven centimetres in length and, as can be seen from the photograph (Fig. 2), is normal in size and conformation in its lower half. A cystic dilatation enlarges the circumference of its upper half. In order that the value of the specimen as a gross exhibit need not be impaired, no incision was made.

The total number of cases observed is, therefore, so far as I can discover, thirty-two including the cases of Treves, Hektoen, and myself. This number indicates the rarity of the disease. Yet I can not help thinking that the innocent character of the lesion has prevented many observers from reporting cases.



Fig. 2.—Professor L. Hektoen's specimen of cystic dilatation of the vermiform appendix.

The symptoms produced by cysts of the appendix are very slight if present at all. In my case there were tenderness and sharp pain on jarring the body for four years prior to the strangulation of the hernia; but the significance of these symptoms can not as yet be determined.

In the absence of characteristic determinative signs the lesion can not be diagnosticated. It will be as much as we can do, as Senn says, to bear this condition in mind when we have to deal with cysts in the region of the appendix. And my own case would cause us to bear in mind the possibility of cystic dilatation when the patient is long afflicted with vague right iliac tenderness aroused into sharp pain by concussion or pressure.

Hernia of the vermiform appendix, uncomplicated by the presence of other visceral parts, is an unusual occurrence. Klein, Brieger, Ba

jardi, and others have collected cases to the number of ninety-eight, to which I may add my own.

Hernias of the vermiform appendix are internal (with which we are not at present concerned) and external. Of the latter, Bajardi found fifty-seven inguinal, forty crural, and one obturator. Considering the relative infrequency of crural hernias, the proportion in this collection seems so great that special causes would seem to operate in its favor. Two circumstances, it seems to me, may be adduced in explanation: First, the small femoral opening might admit an appendix, when a coil of intestine might not enter; and, secondly, in inguinal hernias the greater distensibility of the rings would, as a rule, permit the entrance of other parts of the gastro-intestinal tube.

It is not surprising, in view of the frequency of crural hernias, to find that among eighty-nine cases in which the age was mentioned, thirty-eight were females. The age of the patients is very interesting. In the successive decennia there were respectively fifteen, five, six, five, fourteen, twenty, eleven, three, and one. Of these eighty cases in which the age was mentioned it will be seen that forty-nine cases occurred after the age of forty, and thirty-five cases after the age of fifty years. The explanation, I think, lies in the fact that the lapse of time favors the agencies which bring about descent of the cæcum by elongating the mesocæcum, or relaxing the retrocæcal connective tissue. It is to be noted, at the same time, that twenty-one cases were congenital hernias which were all inguinal ruptures of the right side. Only four of these hernias were free, the remainder being strangulated, inflamed, complicated, or rendered irreducible by incarceration or by adhesions, as occurred in sixteen cases, fixing the appendix to the testis, the spermatic cord, or the wall of the sac. Of the recorded cases, as collected by Bajardi, forty-seven were strangulated. In seventeen of these the appendix was free of adhesions, and ten times it was so little altered that it could be reduced with facility. Eight times the appendix was gangrenous, and three times the constricting band had produced an ulceration. In one case the appendix was reduced without relieving the constriction, the patient dying five hours later.

The clinical phenomena of strangulated appendical hernia are very variable, and have been analyzed by Bajardi for the ninety-eight cases he collected.

The symptoms of antiperistalsis were present in many cases, continuous vomiting occurring fourteen times. Nausea or efforts at

vomiting occurred in five cases. Constipation was absolute in sixteen cases, incomplete in five cases, and wanting in eight instances.

The causes of the symptoms of incarceration are difficult to learn. In Glasmacher's case an acute flexion caused the phenomena, and actual mechanical obstruction doubtless occurs sometimes. A few cases may be explained by Klein's supposition that the obstruction is a dynamic one, due to the reflex involvement of the motor nerves of the intestinal musculature.

Brieger refers in this connection to the experiments of Kirstein, who found that ileus is not produced by the mere sudden occlusion of the intestinal lumen, since the dogs in which intestinal occlusion was brought about by suture died after a rather long time from hunger, while the stormy symptoms which arise soon after the occurrence of incarceration develop as a result of the maltreatment of the strangulated intestine and its nerves. The violent symptoms brought about by the strangulation of omental hernias are also to be borne in mind.

The mechanical conditions upon which hernias of the appendix depend are an abnormally situated cæcum, an unusually lax retrocæcal connective tissue, an exceptionally elongated cæcum, or an excessively lengthy appendix. In the recorded cases the appendix is not said to be extremely long. It would seem that usually the appendix has been able to reach the hernial canal by a postnatal slipping-down process on the part of the cæcum which, in the case of hernias of the cæcum, Hildebrand, who has studied eighty cases of hernia of the cæcum, thinks is the rule. But the occurrence of twenty-one congenital hernias among Bajardi's ninety-eight would lend color to the explanation of Brieger, who states that in fætal life the vermiform process maintains a bandlike connection with the testis, or is immediately adherent to it, so that when the testis descends the appendix is drawn with it. The natural query is, Why does not congenital hernia of the appendix occur oftener if this is correct?

Another ætiological factor suggested by Brieger is the patency of the processus vaginalis. Of course the appendix could easily descend into it upon the occurrence of exciting causes.

Obviously the only explanation of a left-sided inguinal or femoral hernia of the appendix must lie in the abnormal situation of the caput coli, either alone or as part of a general abnormal situation of viscera. As a matter of fact, no case of situs inversus viscerum has yet been observed where the appendix was in a left-sided hernia. The necessary malposition of the cæcum is not unusual.

Inflammation of a herniated appendix is prone to occur, as evi-

denced by Bajardi's thirty cases. The peculiar situation doubtless favors the occurrence of infection, but the presence of foreign bodies in eleven cases accounts for the phenomenon in these instances. Perforation took place in sixteen cases, nine times at the end of the appendix. The appendix was gangrenous in four instances. Diffuse peritonitis occurred seven times as a result of perforation.

The symptoms of hernial appendicitis are those of acute inflammation of greater or less violence in a hernial sac. And if strangulation is added, the symptoms of ileus still further confuse the picture. The diagnosis is very difficult in both strangulation and inflammation of herniated appendices. In forty-seven cases of strangulated hernias of the appendix the diagnosis was, according to Bajardi, only twice made before operation. In Löbker's case Hueter made the diagnosis of probable incarcerated hernia of the appendix on account of the existence of phenomena very similar to those observed in two other cases which he had already seen. In Jackle's case Roser had admitted the possibility of hernia of the appendix on account of absence of vomiting in the presence of intense local symptoms.

Bajardi's comment, that diagnosis of those conditions will be almost always impossible, would seem to be justified when we remember that the symptoms are often so violent as to simulate closely a strangulated hernia of small intestine. In one case a diagnosis of inflamed appendicular hernia was reported by Jackle. A child that had been cured in the clinic of a suppurating inguinal hernia returned presenting a solid cord within the scrotum. The accurate observations previously made rendered the diagnosis easier.

The mistakes made in diagnosis have been varied and sometimes very misleading. The inflamed mass has been at different times considered a peri-orchitis, an orchitis, and an inguinal adenitis, while in one instance the surgeon was in doubt as to the existence of psoitis or coxitis. In the case of Gibney the symptoms closely simulated those of a coxitis.

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103 STATE STREET









